

Hemosuccus pancreaticus

A case report and review of the literature



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Abdullah Ülkü*, Ahmet Gökhan Sarıtaş*, Uğur Topal*, İbrahim Çoğal*, Oğuz Üsküdar**, Atılğan Tolga Akçam*

Cukurova University Faculty of Medicine, Cukurova/Adana, Turkey

*Department of General Surgery

**Department of Gastroenterology

Hemosuccus pancreaticus. A case report and review of the literature

Hemosuccus pancreaticus is a rare clinical condition, defined as a bleeding from an artery around the pancreas or a lesion on the pancreas, to the pancreatic duct. The intensity of bleeding can vary from occult bleeding to life threatening massive acute bleeding. In this article, we aimed to present a 68-year-old man who presented to our clinic with recurrent upper gastrointestinal bleeding and abdominal pain. Endoscopy revealed hemorrhage from ampulla vateri, CT angiographic examination showed an aneurysm of the splenic artery and that the splenic artery was connected to the pancreatic duct. Splenectomy and subtotal pancreatectomy was performed on the patient. The patient was discharged with healing. Hemosuccus pancreaticus should be kept in mind in patients with a history of pancreatitis and peripancreatic aneurysm and upper gastrointestinal bleeding. The early diagnosis and treatment of HP is life-saving. In this study, we aimed to review the basic symptoms and clinical findings, along with the diagnosis and treatment methods of HP.

KEY WORDS: Gastrointestinal bleeding, Pancreas, Splenic Artery

Introduction

Hemosuccus pancreaticus (HP), is defined as gastrointestinal bleeding from the ampulla vateri through the pancreatic duct¹. It is characterized as intermittent bleeding from the ampulla vateri to the duodenum, and is a rare etiology of upper gastrointestinal bleeding. In chronic pancreatitis, aneurysms of the peripancreatic vessels, and pancreatic tumors, an association between the pancreatic duct and the vascular structure arises due to erosion caused by the tumor or pancreatitis. The possibility of the etiology being hemosuccus pancreaticus in cases of upper

gastrointestinal bleeding is estimated to be 1/1500. It is seen 7 times more commonly in men than women, and it usually occurs in the 5th-6th decade^{2,3}.

Lower and Farrell first described main pancreatic duct bleeding in a case with ruptured splenic artery aneurysm in 1931⁴. The term "Hemosuccus pancreaticus" was first used by Sandblom in 1970⁵.

There are a limited number of case reports and reviews in the literature about HP. In this article, we aimed to review the rare case of hemosuccus pancreaticus and its symptoms, diagnosis and treatment in the light of the literature.

Case Report

A 68-year-old male patient with recurrent abdominal pain, hematemesis and melena in the last 1 year was investigated for the etiology of upper gastrointestinal bleeding, the conducted upper gastrointestinal endoscopy revealed active bleeding from ampulla vateri, the CT

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Correspondence to: Uğur Topal, MD, Department of General Surgery, Erciyes University Faculty of Medicine, 38030 Melikgazi/Kayseri, Turkey (e-mail: sutop2005@hotmail.com)

angiography showed a splenic artery aneurysm and the patient was admitted to our clinic with the pre-diagnosis of hemosuccus pancreaticus.

His medical history included hospitalization and medical treatment for GIS bleeding 3 times in the last 1 year. Upper GIS bleeding was accompanied by abdominal pain. Amylase and lipase levels were high in this period. There was hydronephrosis in the right kidney due to essential hypertension and untreated urethral stone. Amlodipine 10 mg was used in the treatment of essential hypertension. He had a history of operation for right kidney stone forty years ago. The patient did not have any contributing family, or psychosocial history. He had no history of smoking or other metabolic diseases. He had no significant alcohol history and no prior clinical or laboratory history of pancreatitis.

On physical examination, his vitals included a blood pressure of 137/81 mm Hg, heart rate was 108 beats per minute, body temperature was 36.8°C, respiratory rate was 19 per minute and oxygen saturation was 99% in room atmosphere. The abdomen was soft and non-tender without organomegaly, other system examinations were normal. The following laboratory results were obtained; WBC: 10.2/mm³, AST: 40 U/L, ALT: 40U/L, ALP:94 U/L, (GGT): 64 U/L, total bilirubin: 0,6m g/dl, direct bilirubin: 0.1-2 mg/dl. Hgb: 11,6 g/dl, Htc 35%, Plt: 144.000 mm³, BUN: 32mg/dl, Creatinine: 0,67mg/dl, sodium: 144mEq/L; potassium: 4.4mEq/L, and albumin: 3.1g/dl.

The most recent endoscopic examination of the patient revealed a clot filling the lumen of the stomach and duodenum, and hemorrhage from the ampulla vateri. Abdominal tomography showed that the right kidney was 29x14 cm in size, a pouch kidney image and aneurysmatic appearance in the splenic artery. CT Angiography

showed an aneurysm of the splenic artery starting from the celiac trunk and associated with the pancreatic duct. It is shown in Fig. 1.

The patient was admitted to the general surgery ward with the diagnosis of Hemosuccus pancreaticus. After anesthetic clearance, laparotomy was performed under general anesthesia. The operation was performed by experienced team of hepatopancreaticobiliary surgeons. In the exploration, the right pouch kidney was aneurysmatic dilated and closely associated with the pancreas, starting from the celiac trunk of the splenic artery. The aneurysm contained a long segment that was too large to be closed by the coil method. After the splenic artery was ligated at the root of the celiac trunk, splenectomy and subtotal pancreatectomy were performed. Because gallbladder was hydropic and wall thickness had increased, cholecystectomy was added. Urology department performed right nephrectomy. The image of the pathological specimen is shown in Figs. 2, 3. Operation time was 3 hours and 15 minutes and blood loss was 250ml. The patient was followed up in the postoperative intensive care unit.



Fig. 1: Splenic artery aneurysm starting from the celiac truncus, in CT angiography.



Fig. 2: Aneurysm in the resection material.



Fig. 3: The nelaton catheter advanced through the pancreatic duct terminates in the aneurysm.

In the postoperative period followed antibiotic therapy (piperacillin-tazobactam), intravenous fluid, analgesics (paracetamol), and anti-emetics were administered, and the oral intake was started after the gastrointestinal passage was achieved. The treatment of the patient, who had no problems, was planned and they were discharged on the 10th postoperative day. The patient had no specific postoperative complications, and a wound complication did not develop. We did not need re-discovery/revision surgeries. We have not experienced postoperative 30-day and long-term morbidity/mortality. The patient was followed up as an outpatient 10 days after discharge and they were well with no further complaints. Another follow-up was done at 3 months.

Discussion

HP is a very rare cause of upper gastrointestinal bleeding. About 150 cases have been reported in the literature since the first report⁶. Cases in the literature are shown in Table I^{2,7-23}. Many causes, such as acute or chronic pancreatitis, pancreatic tumor and vascular malformation, have been implicated as factors affecting HP formation. HP usually occurs as a complication of acute or chronic pancreatitis. Acute pancreatitis causes necrosis of the arterial wall (eg splenic artery, gastroduodenal artery, and duodenopancreatic artery) or enzymatic destruction of the arterial wall, which is effective in the development of pseudoaneurysm and HP occurs after the rupture of the pseudoaneurysm²⁴. Another important cause of HP is pancreatolithiasis secondary to chronic pancreatitis. Intraductal stone compresses the pancreatic tissue to cause necrosis and subsequent vascular ulceration in the wall of the pancreas, causing pseudoaneurysm formation and consequently bleeding^{24,25}. Also, cases of hereditary pancreatitis complicated with HP has been reported in the literature²⁶.

Although pancreatitis is the main factor in the formation of HP, pancreatic tumors can also lead to HP. Primary or metastatic malignant tumors and benign tumors may also result in HP. Pancreatic carcinoma, serous cystic neoplasm, microcystic adenoma, primary pancreatic lymphoma and neuroendocrine tumors are associated with HP. Metastatic cancer, especially renal cell carcinoma (RCC) can cause HP^{24,27-29}.

Vascular anatomy has been detected in approximately 20% of HP cases (24). Atheroma, hereditary dystrophy of elastic tissues (Marfan's disease, Ehlers-Danlos syndrome), fibromuscular dysplasia of arterial walls, portal hypertension, syphilis, vasculitis (especially periarteritis nodosa) and α -1 antitrypsin deficiency are the most common vascular anomalies³⁰.

Most patients with arterial anomalies have arterial aneurysms. Splenic artery is the third most common site of abdominal aneurysms after abdominal aorta and iliac arteries. Primary splenic artery aneurysm is usually

asymptomatic, but may become symptomatic by opening into the pancreatic canal causing HP³¹.

With the development of endoscopic techniques, some cases of HP caused by interventional procedures to the pancreas have been reported. It can be seen as a complication of endoscopic ultrasound-guided fine needle aspiration (EUSFNA) procedure or endoscopic retrograde cholangiopancreatography (ERCP)^{32,33}.

HP cases due to *Brucella* infection and rupture of mycotic splenic artery aneurysm after blunt or penetrating abdominal trauma have also been reported in the literature^{19,34}.

In our case, the etiologic factor in the formation of HP was splenic artery aneurysm.

The trio of epigastric pain, gastrointestinal bleeding and hyperamylasemia are common findings in HP cases, although the clinical presentation is varied. Bleeding may recur within days or weeks after the thrombus within the duct or in the pseudocyst has dissolved. Therefore, HP is characterized by intermittent and recurrent bleeding (24). Clinical signs and symptoms are like upper gastrointestinal hemorrhage manifested by hematemesis and melena. The most important and most common symptom of HP is gastrointestinal bleeding.

Hematemesis is less common than melena. Characteristic colic pain is a result of increased intraductal pressure caused by clot formation in the Wirsung canal. The bleeding is usually not strong enough to cause hemodynamic instability. Chronic anemia symptoms can be seen in many patients because of this reason. In cases of acutely starting massive bleeding, shock can develop in patients³.

Serum amylase levels are often elevated in HP due to increased intraductal/pseudocyst pressure. Other clinical findings include vomiting, weight loss, symptoms of iron deficiency anemia and stool occult blood positivity. Serum bilirubin levels were elevated secondary to pancreatobiliary reflux due to clot obstruction, and liver function tests are normal^{24,32,35}. In our case, there were intermittent abdominal pain, melena and anemia symptoms. He had a history of hospitalization and transfusion due to gastrointestinal bleeding. Amylase, liver function tests and bilirubin were normal.

It is difficult to diagnose HP and it is usually diagnosed late, because the bleeding is intermittent and can heal by itself⁷. Methods such as endoscopy, computerized tomography, CT angiography, MR angiography and angiography can be used for accurate diagnosis. Correctly diagnosing HP in patients presenting with gastrointestinal bleeding is still difficult even with these advanced technological methods.

Endoscopic examination of the upper gastrointestinal tract may indicate bleeding from the pancreas. Endoscopy is the basic method to exclude other causes of gastrointestinal bleeding (erosive gastritis, peptic ulcer, esophagus and other gastric pathologies). Endoscopy can detect active bleeding through the papillae in only 30%

TABLE I - Clinical and laboratory data of twenty-one patients with hemosuccus pancreaticus.

Author	Age/ Sex	Year of Publication	Clinical presentation	Location of Splenic aneurysm	Etiology	Treatment	Pathology
Patel R. ⁷	36/M	2018	Nausea - epi- gastric pain - hematemesis	Proximal splenic artery		Distal pancrea- tectomy + un- block resection	Splenic artery pseudoan- eurysm
Smith J.C. ⁸	24/F	2014	Acute pancrea- titis + melena			Coil emboliza- tion of the pan- creatic duct and duodenum	
Bo li md ⁹	47/M	2017	Hematemesis	Splenic artery aneurysm + pancreatic duct fistula		Splenic artery aneurysm + pancreatic duct fistula	
Wen-Bin Zou ¹⁰	60/F	2015	Abdominal pain + melena (patient oper- ated on due to cholecho- lithiasis and has postop bleeding complaints)		Pancreatic artery opening into the pancreatic duct dorsally	Coil emboliza- tion of the dorsal pancreatic artery	
Hamza A ¹¹	52/M	2017	Abdominal pain + hemato- chezia	Middle splenic artery saccular aneurysm	Splenic artery an- eurysm	Coil Embolization	
Yoshida R ¹²	63/M	2018	Upper abdom- inal pain + hematochezia + temporary loss of conscious- ness	Pseudoaneurysm at SMA	Heterogeneous pseudoaneu- rysm with mural thrombus in SMA + defective area in SMA anterior wall	Coil emboli- zation of the aneurysm after transient hemo- stasis with bal- loon plug +stent implantation after fistula de- velopment after embolization	
Bavanandam S. ¹³	3/M	2016	Recurrent hematochezia -melena + ab- dominal pain + bleeding enough to war- rant transfusion		Leaking at gastro- duodenal artery	2 balloons placed in the gastroduo- denal artery.	
Bavanandam S. ¹³	7/M	2016	Recurrent he- matemesis with abdominal pain + hematochezia		Calcification and dilation at the pancreatic duct	Gastroduodenal Artery Coil Embolization	
Shamah S. ¹⁴	69/M		Upper right quadrant pain + high lipase levels		Arteriovenous fis- tula at pancreatic bed	Embolization	
Maddah G. ¹⁵	33/F	2015	Epigastric pain		Gastroduodenal artery bleeding	Primary suture	
Maddah G. ¹⁵	54/M	2015	Massive gas- trointestinal bleeding		Gastroduodenal artery bleeding	Primary suture	

(continua)

TABLE I - Clinical and laboratory data of twenty-one patients with hemosuccuspancreaticus. (segue)

Author	Age/ Sex	Year of Publication	Clinical presentation	Location of Splenic aneurysm	Etiology	Treatment	Pathology
Kumar N. ¹⁷	50/M	2017	Melena	Superior pancreaticoduodenal artery-posterior inferior pancreaticoduodenal artery	Superior pancreaticoduodenal artery-posterior inferior pancreaticoduodenal artery	Cannulation+ Embolization	
Kothari R.A. ¹⁸	55/M	2013	Ethanol related chronic pancreatitis, recurring melena attacks and abdominal pain	Splenic vein rupture	Rupture of splenic vein into the pancreatic duct	Distal pancreatectomy + splenectomy	Chronic calcific pancreatitis
Kim S.S. ¹⁹	22/M	2000	Firearm injury	Right hepatic artery aneurysm	Right hepatic artery aneurysm	Coil Embolization	
Mandaliya R. ²⁰	61/F	2014	HIV + chronic alcoholic Epigastric pain + melena	Pseudoaneurysm at pancreaticoduodenal artery	Pseudoaneurysm at pancreaticoduodenal artery	Microcoil Embolization	
Ray S. ²¹	46/M	2011	Chronic pancreatitis due to alcohol + necrosis	Splenic artery pseudoaneurysm	Splenic artery pseudoaneurysm	Necrosectomy + pseudoaneurysm ligation	
Ray S. ²¹	26/M	2011	Sudden onset acute abdominal pain after cholecystectomy	Gastrooduodenal artery aneurysm	Gastrooduodenal artery aneurysm rupture	Primary suture	
İnayət F. ²	70/M	2018	Melena	Gastrooduodenal artery aneurysm	Gastrooduodenal artery aneurysm	Embolization	
Koizumi J. ²²	56/F	2000	Abdominal pain ongoing for 1 week and melena + presyncope	Splenic artery aneurysm	Splenic artery aneurysm and fistula between the pancreatic duct	Embolization	
Sul H.R. ²³	48/M	2016	Melena recurring for 6 months	Splenic artery saccular aneurysm	The aneurysm extends from the splenic artery to the pancreas parenchyma	Stent application	

of patients^{24,36}. Since the bleeding due to HO is intermittent, endoscopy may not be enough for definitive diagnosis. Repeated endoscopy may be required to define the problem and a negative endoscopy is not enough to rule out HP^{6,24}.

Ultrasonography can be used to show pancreas pseudocysts or peripancreatic artery aneurysms. Contrasted CT is an ideal method to show pancreas pathologies, it is especially useful in the diagnosis of chronic pancreatitis, pseudocysts and pseudoaneurysms. The presence of coagulated blood in the pancreatic duct known as sentinel clot on pre-contrast CT is a characteristic finding of HP, but is rarely seen³².

CT angiography or selective angiography is the most

effective method for defining peripancreatic aneurysmal disease and diagnosing HP^{37,38}. Selective arteriography of the celiac trunk and superior mesenteric artery provides definitive evidence of hemosuccus pancreaticus by showing passage to the main pancreatic duct. These methods can also determine the existence of another aneurysm or pseudoaneurysm. 96% sensitivity has been reported in selective angiography^{1,37}. However, since bleeding is usually with intermittent episodes, selective angiography may not always localize the source of bleeding. If hemorrhage is localized, angiography is useful not only to find the source, but also to stop bleeding through transcatheter embolization²⁴. Scintigraphy, which is a traditional detection method of bleeding, has

limited use in HP since the symptoms are intermittent. CT angiography and endoscopy were used to diagnose our case. CT angiography showed a connection of the splenic artery aneurysm to the pancreatic duct.

Endovascular minimal invasive procedures or conventional surgical techniques can be used in the treatment of HP. Only supportive treatment is not recommended. The superiority of endovascular treatment over open surgery has not been clearly defined. Open surgical treatment has traditionally included aneurysm resection, splenectomy, and distal pancreatectomy. However, depending on the location of the source of bleeding, the pancreatic resection margin may vary. In endovascular treatments, splenic artery aneurysms are successfully treated with coil or stenting procedures^{7,32}. When the patient becomes hemodynamically stable, invasive methods are effective as initial treatment in 67-100% of the cases. When the source of bleeding is detected on angiography, interventional procedures are the first treatment that can yield good results in 79-100% of cases and the overall success rate is 67%^{1,39,40}.

Bergert et al. recommend a treatment algorithm for HP developing on the basis of pancreatitis and due to arterial pseudoaneurysm. In this algorithm, while angiography and embolization is recommended for hemodynamically stable patients; surgical treatment is recommended for refractory, continuously bleeding patients. Although splenic artery aneurysms can be treated effectively with endovascular treatment, endovascular approach should not be preferred in acute proximal aneurysm. A primary surgical approach is necessary to prevent morbidity in these cases⁴¹.

Surgical treatment is indicated in cases of uncontrolled bleeding, persistent shock, bleeding after first embolization and when embolization is not possible or unsuccessful^{32,42}. Distal pancreatectomy is a surgical alternative for bleeding pancreatic pseudoaneurysms in the body or tail of the pancreas. Surgical resection is associated with increased mortality and morbidity when pseudoaneurysm is localized near the head of the pancreas. The treatment option recommended in these patients is angioembolization^{6,32}. The localization of the source of bleeding, which is an important factor in determining the resection margin of the pancreas, is often difficult. Therefore, intraoperative sonography and pancreatoscopy may be required to confirm the source of bleeding⁴³. Most surgical series reported a success rate of 70% to 85%, mortality between 20% and 25%, and re-bleeding between 0% and 5%^{32,44}. We preferred a surgical approach in our case, and performed a subtotal pancreatectomy along with splenectomy. The localization of the aneurysm was the determining factor in choosing subtotal pancreatectomy instead of distal pancreatectomy. General mortality rate of HP is 9.6%. When treated effectively, recurrence rates are low, except for cases with malignancy. If HP is left untreated, mortality rates can reach 90%^{1,3,24}.

Conclusion

HP is a life threatening and rare cause of gastrointestinal bleeding. HP should be considered in patients with a history of pancreatitis, peripancreatic aneurysm, and upper gastrointestinal bleeding. Although the diagnosis of HP is difficult due to the intermittent bleeding pattern, the diagnosis can be confirmed by selective angiography. The diagnosis is confirmed in all patients by means of examinations, preferably during active bleeding. Appropriate diagnostic studies and treatment methods are important in reducing the mortality rate. Angioembolization can be performed in the treatment and surgical treatment can be applied if it fails. However, urgent surgical intervention is required in patients with recurrent bleeding or failure of embolization. Gastroenterologists and surgeons should be familiar with this disease, because early diagnosis can be lifesaving.

Riassunto

L'emosuccus pancreatico – o wirsugorragia - è una rara condizione clinica, definita come sanguinamento da un'arteria del pancreas o da una lesione sul pancreas nel dotto pancreatico. L'intensità del sanguinamento può variare da sanguinamento occulto a sanguinamento acuto massiccio potenzialmente letale. In questo articolo, presentiamo il caso di un uomo di 68 anni che presentava ricorrenti sintomi di sanguinamento del tratto digestivo superiore e dolore addominale. L'endoscopia ha localizzato l'emorragia dall'ampulla Vateri, l'esame angio-TC ha rivelato la presenza di un aneurisma dell'arteria splenica adiacente al dotto pancreatico. Splenectomia e pancreatectomia subtotale sono state eseguite sul paziente ed il paziente è stato dimesso guarito. L'emosuccus pancreatico deve essere tenuto presente nei pazienti con anamnesi di pancreatite e aneurisma peripancreatico e sanguinamento del tratto gastrointestinale superiore. La diagnosi precoce e il trattamento dell'HP sono salvavita. In questo studio, abbiamo sottolineato i sintomi di base e i risultati clinici, insieme ai metodi di diagnosi e trattamento del HP.

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Commento e Commentary

PROF. NICOLA PICARDI
Già Ordinario di Chirurgia Generale

Il caso presentato è certamente interessante nella sua relativa rarità, anche se l'emorragia emergente dal dotto del Wirsung – nota anche come “wirsungorragia” – è una eventualità conosciuta quale raro segno indiretto di patologia pancreatitica, neoplastica o vascolare.

Rappresentava certamente una rarità degna di citazione nelle casistiche cliniche per le indubbie difficoltà diagnostiche prima che la moderna semeiotica strumentale ne rendesse facile e immediata la diagnosi. Ad ogni eventualità ricorrente di melena ed ematemesi il protocollo da seguire si fonda su due pilastri: l'endoscopia digestiva e l'angio-Tac. Con la duodenoscopia è facile individuare la fonte dell'emorragia dalla papilla di Vater, ponendo la diagnosi differenziale tra wirsungorragia ed emobilia, e la storia clinica di un pregresso trauma può aiutare. L'angiotac, in grado di identificare patologia vascolare o flogistica o neoplastica in ambito pancreatico o epatobiliare, permette la diagnosi etiologica dell'emorragia, guidando le scelte terapeutiche: embolizzazioni angioradiologiche conservative quando possibili o demolitive chirurgiche, più relativamente più facili se la patologia causa dell'emorragia è a sede pancreatica, molto più problematica se a partenza epatobiliare.

Anche l'angiografia selettiva del tripode celiaco e dell'a. mesenterica superiore, ha un suo ruolo nella ricerca diagnostica, purché eseguita in corso di emorragia e di flusso adeguato.

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The presented case is certainly interesting in its relative rarity, even if the hemorrhage emerging from the duct of Wirsung – also known as “wirsungorragia” – is an eventuality known as a rare indirect sign of pancreatitic, neoplastic or vascular pathology of the pancreas.

It certainly represented a rarity worthy of mention in clinical case studies due to the undoubted diagnostic difficulties before the modern instrumental semeiotics made its diagnosis easy and immediate. For each recurrent eventuality of melena and hematemesis the protocol to follow is based on two pillars: digestive endoscopy and angio-Tac. With duodenoscopy it is easy to identify the source of hemorrhage from Vater's papilla, placing the differential diagnosis between wirsungorragia and hemobilia, and the clinical history of a previous trauma can help.

The angiotac, able to identify vascular or phlogistic or neoplastic pathology in the pancreatic or hepatobiliary environment, allows the etiological diagnosis of hemorrhage, guiding the therapeutic choices: conservative angioradiological embolizations when possible or surgical demolition, more relatively easier if the pathology causes of the hemorrhage is at the pancreatic site, much more problematic if at the beginning hepatobiliary.

Also the selective angiographies of the celiac tripod and superior mesenteric, have a role in diagnostic research, provided performed during bleeding with adequate flow.